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Case Report

Dermatologic manifestations of complex regional pain syndrome improved after dorsal root ganglion stimulation

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ABSTRACT

Background: Complex regional pain syndrome is a condition that causes autonomic dysfunction, including severe pain, swelling, temperature fluctuations, and cutaneous flushing.

Case Description: The patient was a 38-year-old woman with complex regional pain syndrome type I of the right foot that developed after a work-related accident. At the time of presentation, she complained of a stabbing and burning pain, which subjectively felt like extreme warmth for 6 weeks. On physical examination, she presented with diffuse cutaneous flushing, erythema, nonpitting edema, skin mottling of the medial aspect of the right foot and ankle, a purple macule on the right inferomedial aspect of the great toe, and a purple patch on the inferomedial aspect of the heel. The patient underwent right-sided L5/S1 open dorsal root ganglion (DRG) stimulation under the care of neurological surgery. Postprocedurally, the cutaneous flushing, erythema, edema, mottling, purple macule, and patch had completely resolved. At the 1-year postoperative appointment, the patient continued to have a significant improvement in her preoperative pain and notably improved allodynia, flushing, mottling, and temperature sensitivity.

Conclusion: We have described the successful resolution of complex regional pain syndrome associated dermatologic manifestations in the setting of DRG stimulation. To our knowledge, a case of this nature has yet to be described in the literature.

Keywords: Chronic pain/diagnosis, Chronic pain/therapy, Complex regional pain syndromes*/therapy, Ganglia, spinal/physiology

INTRODUCTION

Complex regional pain syndrome (CRPS) is a condition that causes autonomic dysfunction, including severe pain, swelling, temperature fluctuations, and cutaneous flushing. CRPS is categorized into types I and II as defined by the Budapest criteria; type I has an underlying etiology of nerve injury or illness, and type II is related to damage of a specific nerve.[8] This syndrome has been associated with injuries, such as fractures, sprains, and prior surgical intervention,[15,16] but there is no widely agreed upon mechanism for CRPS.[5] While the exact mechanism for the condition remains unclear, CRPS is believed to be a result of central and peripheral nervous system sensitivity changes with an increase in inflammation and dysregulation of the autonomic

nervous system. Patients with CRPS have higher levels of inflammatory molecules, including interleukin-6, interleukin 1-beta, and tumor necrosis factor-alpha, which may trigger the release of histamine and lead to induration, redness, warmth, and pain.[5,10] Cutaneous manifestations of CRPS may also result in dermatitis, folliculitis, bullae, erythematous papules, and mild ulcerations.[5,9-11]

There are many pharmacological and minimally invasive treatments for CRPS that have varying efficacy. For example, gabapentin, nonsteroidal anti-inflammatory drugs, corticosteroids, ketamine infusions, and even bisphosphonates have been used to treat CRPS, and sympathetic nerve blocks have also shown positive results for pain reduction.[15,16] However, some patients experience refractory symptoms in response to these therapies. In these cases, procedural intervention may be considered. For procedural options, spinal cord stimulation (SCS) and dorsal root ganglion (DRG) stimulation have been shown to be safe and effective means of treating these patients.[12,16]

Prior attempts at leveraging invasive neuromodulation techniques in CRPS patients predominately targeted the spinal cord, but there is foundational evidence suggesting that DRG stimulation is a potentially more effective approach than other methods.[1] From a neurophysiologic perspective, DRG stimulation is a more targeted approach than SCS, as DRG stimulators act peripherally versus centrally, as in SCS. In addition, functional improvements in neuropathic pain after SCS have been inconsistent.[1] Animal models of chronic pain have targeted the DRG to mediate neuronal hyper-excitability in nociceptive pathways.[1] Therefore, the DRG can be a viable target for neuromodulation. DRG stimulation was approved to treat refractory lower limb CRPS in 2016.[6] To our knowledge, there are no published reports that visually demonstrate the cutaneous improvements in CRPS with DRG stimulation.

CASE PRESENTATION

The patient was a 38-year-old woman with a past medical history of CRPS type I of the right foot after a work-related incident. She was injured when a heavy object landed on her foot and caused a closed, nondisplaced fracture of the anterior process of the right calcaneus and a sprain of the talofibular ligament and calcaneofibular ligament of the right ankle in November of 2022. After this accident, she presented to the neurosurgery clinic in January of 2023 complaining of pain, which was burning and stabbing in quality, with excessive skin warmth for the past 6 weeks. Physical examination revealed nonpitting edema, erythema, cutaneous flushing, and skin mottling localized to the medial aspect of the foot and ankle and purple macule and patch on the inferomedial aspect of the right foot and ankle, respectively. The initial differential diagnosis included peripheral vascular disease, Dorsal root ganglion stimulation for dermatologic manifestations of complex regional pain syndrome, peripheral nerve impingement, tenosynovitis, and Charcot's foot. Vascular surgery was consulted, and performed a comprehensive examination of pulses reported normal ankle-brachial-indices and confidently ruled out vascular etiology. Given the quality of the patient's pain, temperature sensitivity, and cutaneous findings, we were primarily concerned with a primary neurologic versus vascular etiology. In the context of the vascular examination and her presentation, which fulfilled the Budapest criteria (allodynia, skin color changes, edema, temperature sensitivity, and range of motion limitations), we ultimately favored a primary neurologic etiology versus vascular etiology, eventually diagnosing her with CRPS type I [Figures 1 and 2].[8]

She underwent a percutaneous DRG stimulation trial under the care of pain management with approximately 80%

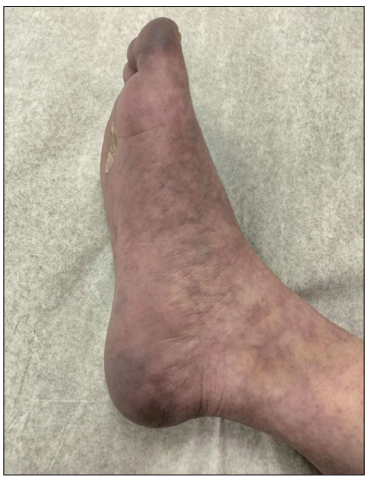

Figure 1: Skin mottling and darkening from complex regional pain syndrome-1 of right foot and ankle.

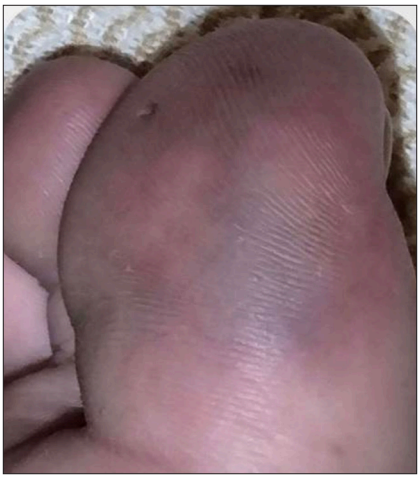

Figure 2: Magnified view of right hallux longus on the right foot.

improvement in her pain at 2 weeks of clinical follow-up. In addition, there was complete resolution of the right foot and ankle edema, cutaneous flushing, erythema, mottling, hyperpigmented macule on the great toe, and hyperpigmented patch on the heel at 2 weeks of clinical follow-up. Because of this outcome, she was recommended for a right-sided L5/S1 laminectomy with DRG stimulator placement under the care of neurological surgery. At the 1-year postprocedure appointment, the patient continued to have significant improvement in her preprocedure pain and notably improved allodynia, flushing, mottling, and temperature sensitivity [Figures 3 and 4].

DISCUSSION

In this case, the patient's pain and skin changes improved (detailed description in case presentation) after DRG stimulator placement, providing more evidence of the efficacy of DRG stimulation in treating CRPS and its cutaneous manifestations. Dermatologic manifestations of CRPS can be difficult to identify, given a general nonspecificity of findings. Because of this, descriptions of this entity are limited across the literature. Given that skin changes are a component of the Budapest criteria, a thorough understanding of their manifestation and dynamics is necessary for a better understanding of CRPS overall. Furthermore, dermatologic manifestations of the CRPS disease process are present in all three of the defined stages, which was reinforced by the presence of skin changes from the outset in our described case.[14] Previous literature has found that vascular manifestations are the most frequent skin change noted in CRPS.[14] Slightly more than half of all patients with CRPS present with edema and erythema, both of which were present in our presented case.[14] Additional findings are dermatitis (35%), erythematous papules (23%), and cutaneous atrophy (23%).[14] Furthermore, our case presented two hyperpigmented skin changes, a feature present in approximately 8% of CRPS cases.[14] Traditional management of CRPS-related dermatologic manifestations is difficult and typically requires control of the underlying CRPS disease process.[14] Contemporary management includes nerve blocks, local anesthesia, physical/occupational therapy, and SCS.[13,17] However, a formal description of the effects of these therapies, and particularly DRG stimulation, on the resolution of CRPS-associated dermatologic manifestations has not been to our knowledge described in the literature.

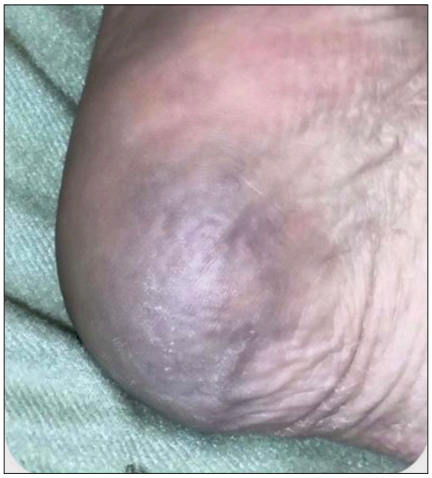

Figure 3: Magnified view of right heel.

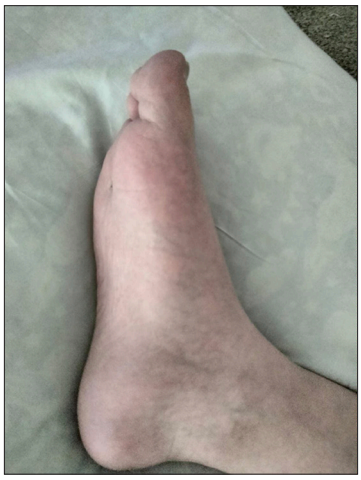

Figure 4: Improvement in cutaneous symptoms.

DRG stimulation has more advantages than dorsal column SCS because it is a more targeted therapy for the affected pseudo-unipolar afferent nerve fibers.[2] The ACCURATE trial previously demonstrated the efficacy of DRG stimulators compared to SCS in patients with persistent type I or II CRPS.[4] At 3- and 12-month post-DRG stimulator or -SCS placement, patients with DRG stimulators more frequently met the criteria for treatment success (>50% pain reduction) than patients with a SCS.[4] Similarly, a small prospective study by Vallejo *et al*. found that nine patients who completed the 6-month follow-up were all satisfied with their improvements to some degree, and most of them were at least moderately satisfied with the results of the procedure.[18]

DRG stimulation has been studied within the context of both upper and lower-extremity CRPS. In one of the first prospective series to examine the use of the DRG as a target for neurostimulation in the context of CRPS, Van Buyten *et al*. recruited eleven patients with lower extremity (foot and leg) CRPS, representing a variety of etiologies.[19] DRG stimulators were placed at various lumbar DRG locations and trialed, with eight out of eleven patients ultimately receiving permanent stimulator placement. At 1 month, there was a 62% reported reduction in pain levels compared to baseline values. In addition, pain relief was durable in the majority

of patients (75%).[19] Graca *et al*. evaluated DRG stimulation in the cervical and high thoracic spine of patients diagnosed with CRPS I or II that was refractory to prior treatment.[7] After an initial stimulation trial, 85% of patients reported successful DRG stimulation therapy (≥50% reduction in pain defined by a numeric rating scale) with durable pain relief in 6 months.[7] Because of the promising pain reduction outcomes, there is a greater shift toward utilizing DRG over SCS in cases of CRPS. Therefore, as the field shifts in this direction, there ought to be a greater effort to describe the effects of DRG stimulation on the less historically characterized manifestations like skin changes. Our case provides preliminary evidence for efficacy in managing dermatologic CRPS manifestations with DRG stimulation.

While there are some reports of SCS improving dermatologic manifestations of CRPS, there is still limited literature documenting the resolution or improvement of these cutaneous symptoms after surgical intervention.[3] Dermatologic symptoms are postulated to occur in an acute and chronic phase of CRPS. The acute phase of the condition involves a sudden influx of norepinephrine due to a decrease in sympathetic nervous system activity. Due to the increase in available norepinephrine, there is an upregulation and sensitization of peripheral α-1 adrenergic receptors, causing vasodilation.[15,16] The chronic phase of CRPS is characterized by a decrease in peripheral α-1 adrenergic receptors, causing vasoconstriction.[15,16] Crapanzano *et al*. reported a case of a 53-year-old female with CRPS type I whose pain, erythema, swelling, and tissue necrosis significantly improved after SCS placement.[3] Our case report highlights the efficacy of using DRG stimulation rather than SCS for CRPS and the additional benefit of resolving cutaneous manifestations.

CONCLUSION

Herein, we describe a case of CRPS 1 with prominent cutaneous physical exam findings. A DRG stimulator was placed with a 2-week clinical follow-up. At this time, not only did the patient experience a reduction of pain but also there was also resolution of her skin changes. Given these findings, DRG stimulation may have the ability to manage complex regional pain syndrome-associated dermatologic manifestations effectively. Historical approaches to complex regional pain syndrome, such as SCS, nerve blocks, and physical therapy, are limited in their emphasis on the ability to manage skin changes. The take-home message of our report is such that we have potentially provided an alternative approach to managing complex regional pain syndrome-associated dermatologic manifestations but are limited in our discussion by a scarcity of similar literature.

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